Recurring Peripheral Ameloblastoma at Mandibular Premolar Region: A Case Report

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Abstract

Peripheral ameloblastoma is an extraosseus type, rare form of ameloblastoma that proliferates on the soft tissue of tooth bearing region. It is usually an exophytic odontogenic tumour that exhibits with either smooth or irregular surface and is mainly located in the mandibular region namely the gingival area. This lesion is mainly limited by the periosteum but a larger lesion can have features of bony marginal saucerization as well as displacement of teeth. We hereby report a case of recurring peripheral ameloblastoma on the mandibular left premolar region in a 37 year-old Malay gentleman seen at the Kulliyyah of Dentistry, International Islamic University Malaysia.

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Introduction

Ameloblastoma is ranged to be 1% of all oral tumour and also 11% of all types of odontogenic tumour.[1] The World Health Organization (WHO) 2005 histological classification of odontogenic neoplasms; describes ameloblastoma as a benign yet locally invasive epithelial odontogenic tumour of enamel organ origin.[2] Ameloblastoma is postulated to arise from either the remnants of dental lamina, epithelial lining of an odontogenic cyst, or from the basal cells of the oral mucosa.[3] In general, ameloblastoma is classified as conventional intraosseus form; categorized further as solid, multicystic or unicystic while the extraosseus ameloblastoma is an untypical form. Male to female ratio of conventional ameloblastoma occurrence is 2:1 with more predilection towards mandible as compared to maxilla.[4] Histologically its feature appears similar to that of cutaneous basal cell carcinoma,[5] while it can be well differentiated from peripheral odontogenic fibroma.[6] Peripheral ameloblastoma which is an extraosseus form of ameloblastoma comprises about 2-10% of all known occurrence of ameloblastoma.[7]It is an exophytic growth that occurs on the soft tissue near the tooth-bearing areas of the jaw.[8]

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Case Report

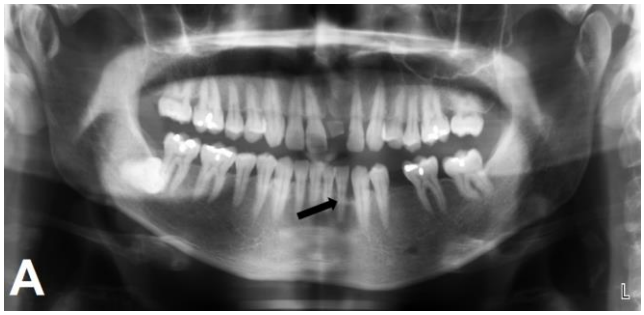
In 2012, a 37 years-old Malay gentleman attended the Oral Pathology and Oral Medicine Diagnostic Polyclinic at the Faculty of Dentistry, International Islamic University Malaysia, Kuantan, Malaysia with the complain of a painless, slow growing mass on the lower left premolar region. The asymptomatic swelling is firm yet sessile mass, measuring 2 cm by 1.2cm in dimension with finger-like surface projections located on the lingual region of 31 to 34 teeth area (Figure 1).

The surrounding tissue appeared normal and healthy with no signs of cervical or submandibular lymphadenopathy. Incidentally, the teeth 31, 32, 33 and 34 were vital and patient had no relevant medical history. Additionally, the Orthopantomograph as well as intra-oral periapical dental radiographs showed an interdental spacing between 33 and 32 with minimal horizontal bone loss and having no clear signs of aggressive bone erosion (Figure 2).

Differential diagnosis of pyogenic granuloma, gingival epulis or peripheral giant cell

granuloma was made. The entire lesion was biopsied in total under local anaesthesia. The excised soft tissue was sent for histopathological examination at the Department of Pathology, Hospital Tengku Ampuan Afzan, Kuantan, Pahang.

Figure 1. Intra-oral Exophytic Swelling on the Anterior Mandibular Region Seen in 2011.



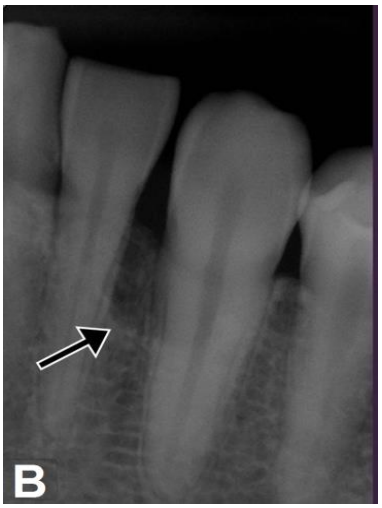


Figure 2. Pre-operative radiograph shows interdental spacing and horizontal bone loss at region between the teeth 32 and 33 (see arrow). (A) Orthopantomograph and (B) Intra-oral periapical.

Specimen's Hematoxylin and Eosin (H&E) stained histology slide (Figure 3) showed islands of odontogenic epithelium composed of loosely arranged stellate reticulum like cells surrounded by palisaded columnar ameloblast-like cells with reverse polarity. There is also squamous metaplasia with keratinization in the stellate reticulum like cells; while some areas show extensive metaplastic squamous cells with prominent nuclei. The section is covered by parakeratinised stratified squamous epithelium. In view of the clinical, physical and radiographic features; a preliminary diagnosis of peripheral ameloblastoma was made. However, the patient failed to appear for review in follow-up appointments.

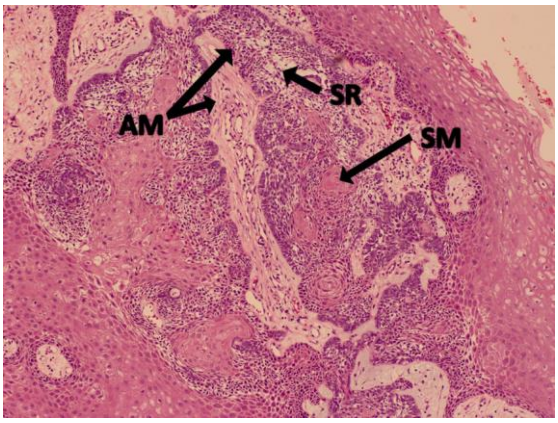


Figure 3. Histological Features of Biopsied Specimen. AM: *Ameloblast-like cells*, SR: *Stellate Reticulum*, SM: *Squamous Metaplasia*.

Figure 4. Recurrence of the exophytic mass in the same anterior mandibular region in 2014.

Eventually, in 2014 the patient had returned with a recurrence of swelling on the lingual and labial gingiva at the same site as seen in the year 2011 (Figure 4). The swelling appeared similar to its past presentation. This time however, the

mass has extended onto the buccal aspect with the teeth 32 and 33 being mobile and non-vital.

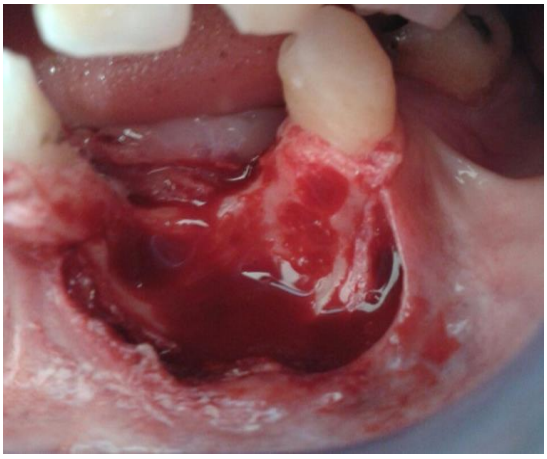


Figure 5. Feature of Bony Saucerization, with No Penetration Tissue Tags during Excision.

Repeated biopsy yielded similar histopathological feature as before. Following discussion with patient, a total tumour resection with 5 mm clear margin peripheral ostectomy as well as extraction of 32 and 33 was done. Upon removal of tumour and extraction of 32 and 33, the underlying bony bed showed clear saucerization with no penetrating tissue tags (Figure 5). The exposed bone surface was covered by advancement flap from the labial region of teeth 31 to 34. The operation site healed without any complications. Post-operative annual clinical and radiographic review did not show any signs of recurrence.

Discussion

Peripheral ameloblastoma is a very rare type of odontogenic tumour to be encountered.[9,10] Although peripheral ameloblastoma commonly occurs in patients aged more than 50 years old, there is documentation stating that patients could range from 20 up to 80 years old.[11] The patient depicted in this case study was a 37 years old gentleman who is well within the age range of this lesion occurrence. Interestingly, premolar region is a common occurrence site according to previous cited report which is in agreement with our case.[11] Though classically known to have a smooth surface; this tumour can exhibit granular, papillary, pebbly or warty surface.[3] This feature was clearly seen in our case study. Feature of marginal saucerization was seen both radiologically and intraoperatively, which was reported in prior reviews.[12] Additionally, peripheral ameloblastoma lesions that are located in an interdental papilla area may lead to teeth displacement,[5] a feature consistent with our case study. Despite the fact that peripheral ameloblastoma prerequisite the exclusion of bony extension, histologically speaking it is a tumour with ability to extend into other tissue plane including bone. Hence, the occasional rare case of intraosseus lesions as being peripheral ameloblastoma should not dismissed completely as cited in a case report.[13]

Peripheral ameloblastoma's diagnosis usually stands apart distinctively based on its histologic evaluation.[11] This lesion is mainly composed of follicular odontogenic epithelial cell islands that can be presented as acanthomatous, with areas of centralized keratin formation.[14] The epithelial cell islands or strands are usually surrounded by fibrous tissue.[15] These histological features mimics infallibly to that seen in this case study. Most peripheral ameloblastoma lesions are surgically dealt with via local excision with a minimal margin of normal tissue. The tumour is usually limited at its base by the periosteum which is usually included in the excision in order to avoid bone infiltration.[9,16] It is common believe that unlike the usual ameloblastoma tumours, peripheral ameloblastoma does not show features like invasiveness as well as uninhibited growth,[17] however this is not always a definite feature. As for our patient in this case study, local excision with a minimal 5 mm margin and minimal peripheral ostectomy over underlying bone surface was done once histological results show no features of invasiveness. Post-surgical follow-up review showed no features of recurrence too.

Conclusions

In conclusion, common literature describes peripheral ameloblastoma as being a rare lesion. Despite being benign and slow growing, its constant nature will eventually cause the tumour's growth to such large proportion that it could affect oral function such as mastication, speech, aesthetic and even breathing. Given enough time, the lesion could be inevitably traumatized, that may lead to greater complications. A definitive management is required to avoid such morbidity to befall the patient.

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Declaration of Interest

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